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ORIGINAL ARTICLE

Lateral meningocele with asymmetric canal stenosis: A case study

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Abstract Lateral meningocele is a very rare disorder of unknown aetiology typified by the presence of protrusions of the arachnoid and the dura matter extending laterally through inter- or intra-vertebral foramina. We report here the case of a 52-year old male with abnormality of spine when presented with low back pain. The patient did not appear to have any neurological disorder. A computerized tomography (CT) scan was acquired from T12 to mid sacrum with multiplanar reformations. The results showed the presence of a left sided paraspinal cystic lesion projecting from the left neural foramen and extending into the left psoas muscle suggesting a lateral meningocele. In addition, a broad based central and left paracentral disc protrusion was also observed resulting in asymmetric canal stenosis. The patient is on regular follow-up while undergoing palliative treatment.

1. Introduction

Lateral meningocele syndrome is a rare disorder characterized by the widespread presence of protrusions of the arachnoid and the dura matter extending laterally through inter- or intra-vertebral foramina into the paraspinal, retroperitoneal, or intrathoracic region (Oner et al., 2004). It is a hereditary disorder that primarily affects the connective tissues. This disorder manifests itself with formations of cysts at different levels of the central nervous system along with meningeal diverticula protruding through the intervertebral spaces and

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filled by cerebrospinal fluid (Nabors et al., 1988). Other clinical findings associated with the lateral meningocele syndrome include specific facial anomalies, cryptorchidism, hypotonia and muscle atrophy, scoliosis, restricted joint movements, pectus deformities, and abdominal hernias (Lehman et al., 1977; Chen et al., 2005). Lateral meningoceles may be unilateral (as in the present case) or bilateral and may exist as solitary (as in the present case) or multiple. The pathogenesis of lateral meningoceles is complex because symptom onset depends on the location of the cyst and the meningeal protrusions. So far, lateral meningoceles have been reported in less than 20 patients worldwide, which include two cases of dominant genetic transmission while a few cases of sporadic occurrence in males and females (Nabors et al., 1988; Lehman et al., 1977; Katz et al., 1978; Philip et al., 1995; Gripp et al., 1997; Alves et al., 2013). Presently, genetic basis and phenotypic variability in lateral meningoceles patients are poorly understood. In the current clinical report, we will discuss the case of a 52-year old male who had lateral meningoceles with no evidence of associated craniofacial abnormalities, Marfan's syndrome or neurological disorder.

2. Clinical report

This 52-year old male patient came to our attention due to the presence of spinal meningeal protrusions. At 47 years of age, he began to suffer from recurrent and chronic back pain with associated neuralgia in the lower extremities, most likely due to irritation of the sensory root of spinal nerve(s). Higher mental functions and motor and sensory systems of the patient appeared normal upon neurological exam. No neurological deficits were noted. The patient's stature was normal and skin lesions were absent. Bladder and bowel functions were functioning normally. Past history included chronic pain of musculoskeletal origin which was more pronounced at the lower back and the knees, post-exertional malaise, easy fatigability, and chronic headache without aura and occipital neuralgia.

A computerized tomography scan (CT scan) of the lumbosacral spine of the patient was ordered since CT scan is the diagnostic test of choice to study spine disorders. CT imaging was acquired from T12 to mid sacrum with multiplanar reformations (Figs. 1 and 2). CT scan revealed the presence of bilateral partial sacralization of the L5 vertebra with anomalous articulations between the L5 transverse process and the sacrum bilaterally. Vertebral body alignment and disc space heights were found to be within normal limits with no spondylolisthesis or pars defects. Facet joints outlined normally. There was no evidence of a recent fracture. At the L1/2 level, a focal right far lateral disc osteophyte complex was noticed which was adjacent to the right L1 nerve root as it exited the neural foramen. On the left side at the L1/2 level, there was a large well circumscribed paraspinal cystic lesion lying within the left psoas muscle which appeared continuous with the left L1 neural foramen. This cystic lesion measured 61 mm (millimetres) in craniocaudal length by 43 mm in width and 33 mm in anteroposterior diameter. This was felt most likely to represent a lateral meningocele (Fig. 2). Furthermore, at the L2/3 level there was a broad based left foraminal and far lateral disc osteophyte complex abutting the exiting left L2 nerve root. At the L3/4 level, there was a broad based central and left paracentral disc protrusion resulting in moderate asymmetric canal stenosis and potentially impinging upon the left L4 nerve root as it entered the lateral recess. Likewise, at the L4/5 level there was a broad based central posterior disc osteophyte complex resulting in mild canal stenosis. These findings were suggesting of lateral meningocele. In order to prevent the patient's daily activities from getting worse, analgesics were used for the patient's pain management along with weekly physical therapy and follow-up visits with the physician.

3. Discussion

In our patient, milder manifestation of clinical symptoms of lateral meningocele allowed him to remain undiagnosed for years before the start of chronic back pain due to spinal root and vertebral compression. Surgical correction of the meningeal protrusion at a later stage of diagnosis is not the best option due to the likelihood of significant worsening of disability with irreversible nerve damage. Castori et al. have reported the case of a 55-year old woman with the lateral meningocele syndrome who underwent surgery to correct two large lateral meningoceles at the lumbosacral level which were supposedly

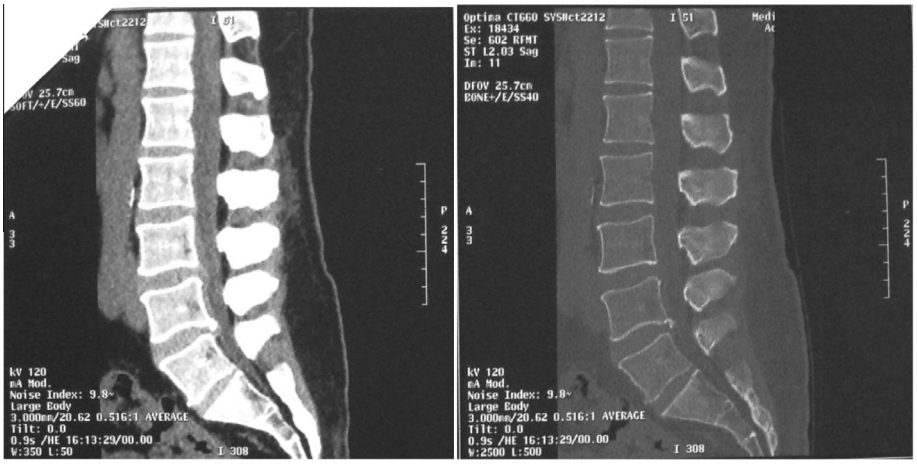

Figure 1 Serial sagittal CT (computerized tomography) scan images of dorso-lumbo-sacral spine showing the presence of lateral meningoceles.

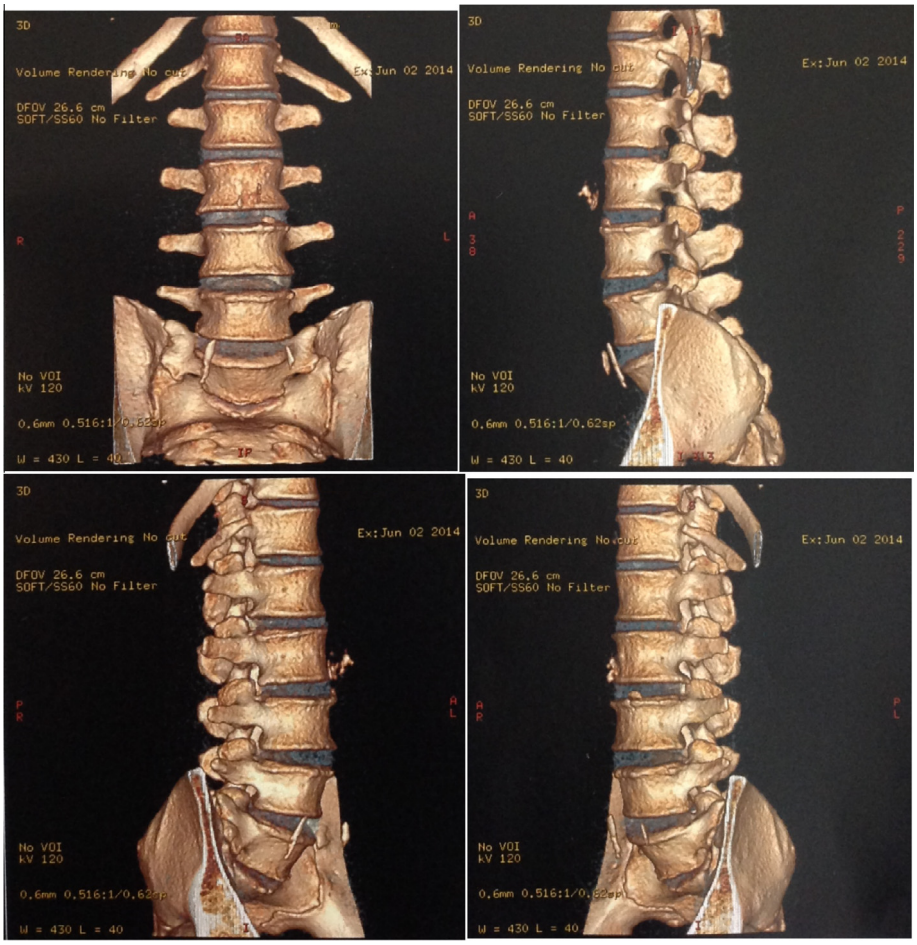

Figure 2 Colored 3D computed tomography (CT) scans of the lumbo-sacral spine with lateral meningoceles showing multiple frontal and side views. Osteophyte formation can be seen at the L1/2 level, adjacent to the right L1 nerve root as it exits the neural foramen. On the left side at the L1/2 level, a large well circumscribed paraspinal cystic lesion lying within the left psoas muscle is present which appears continuous with the left L1 neural foramen representing a lateral meningocele. Furthermore, a broad based left foraminal and far lateral disc osteophyte complex abutting the exiting left L2 nerve root is evident at the L2/3 level. At the L3/4 level, there was a broad based central and left paracentral disc protrusion resulting in moderate asymmetric canal stenosis.

the cause of her pain. However, surgery did not alleviate her symptoms and only exacerbated the symptoms by causing irreversible nerve damage with bladder and anorectal dysfunction linked to weakness in lower limbs, hence making the patient wheelchair bound (Castori et al., 2014). Similar unforeseen complications emerged when a 40-year old woman with the lateral meningocele syndrome underwent surgical correction of the meningeal protrusions (Isono et al., 1999). This accentuates the importance of early diagnosis of the lateral meningocele syndrome in case that continuous pain is presumed to originate from a lateral meningocele, and surgical removal is considered at an early stage to avoid complications (Castori et al., 2014). Likewise, surgical excision may be indicated in cases where giant and symptomatic cysts are present and they are causing bladder or bowel dysfunction or neurological deficits (Kim et al., 2011; Gocer et al., 1999).

A patient with lateral meningoceles may remain asymptomatic or may suffer from paraparesis or back pain (Mizuno et al., 2002). Chronic symptoms in lateral meningoceles usually arise when meningeal protrusions compress against or deform the adjacent structures such as vertebral bodies, nerves, and viscera. Generally, lateral meningoceles are present in the form of occult lesions that are not visible externally. One factor that has been suggested to contribute to the formation of lateral spinal meningoceles is the imbalance between both the pulsations of the cerebrospinal fluid and its hydrostatic pressure, and the resistance of the dura mater and the arachnoid to distortion by such pressure, particularly at the intervertebral foramina. Hence, the arachnoid and the dura may project outward through it in case of a developmental body defect (Panil Kumar et al., 2013). Lateral meningoceles occur more commonly in females than males. Clinical signs of lateral meningoceles syndrome usually manifest in patients when they are in their forties or fifties although younger patients have also been reported (Seddighi and Seddighi, 2010).

Our patient is one of the oldest reported individuals with lateral meningoceles. The diagnosis was established on the basis of CT scan of the lumbosacral spine. Lateral meningoceles appear on CT scan in the form of well-defined, homogeneous, low-attenuation paravertebral masses (Webb, 1994). Radiographic studies (Figs. 1 and 2) revealed the presence of a large, left sided paraspinal cystic lesion extending into the left

psoas muscle measuring up to 61 mm in maximum diameter. The cyst appeared to project from the left L1 neural foramen and was felt most likely to represent a lateral meningocele. This can be further confirmed with a magnetic resonance imaging (MRI) examination because of the ability of MRI scans to further describe the details of multiple lesions such as paravertebral expansion and dural ectasia (Reis et al., 2005). On the right side at the L1/2 level, there was a focal far lateral disc osteophyte complex abutting the exiting right L1 nerve root. There was a broad based left foraminal and far lateral disc osteophyte complex at the L2/3 level abutting the exiting left L2 nerve root. A broad based central and left paracentral disc protrusion at the L3/4 level appeared to result in moderate asymmetric canal stenosis, thereby potentially impinging upon the left L4 nerve root as it entered the lateral recess. A broad based central disc osteophyte complex at the L4/5 level was found to result in mild canal stenosis. To sum up, this rare finding may be beneficial for radiologists and surgeons. Further clinical and molecular studies (including MRI scans) need to be conducted in order to find the genetic aetiology of lateral meningoceles in this patient.

Conflict of interest

The authors confirm that this article content has no conflict of interest.

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